

VAGINAL OBLITERATION IN A WOMAN WITH A HISTORY OF CUTANEOUS T-CELL LYMPHOMA: THE RESULTS OF COMBINED CHEMOTHERAPY-INDUCED GONADAL TOXICITY AND LYMPHOMA RELAPSE

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SUMMARY

Objective: Although ovarian failure commonly occurs following chemotherapy, the relation between menopause or related sexual activity in this failure is often overlooked. We report a case that emphasizes a clinically rare complication, the complete obliteration of the vagina.

Case Report: A 42-year-old married woman with a history of Mayer-Rokitansky-Küster-Hauser syndrome, an established neovagina, and complete clinical remission of cutaneous T-cell lymphoma (stage T4) treated with bleomycin, cyclophosphamide, doxorubicin, vincristine and prednisolone had suffered from abdominal pain. Imaging studies (computed tomography) and laboratory evaluations indicating a suspicious pelvic abscess with leukocytosis (16,800/mm³) and elevated serum C-reactive protein (18.4 mg/L) levels. The patient underwent intensive medical treatment and surgical intervention, but died 2 months later. Pathology showed widespread lymphoma throughout the perineal area, including the neovagina, deep pelvic cavity and the entire abdominal cavity.

Conclusion: Physicians should be greatly concerned about the frequency and severity of treatment-associated acute and long-term complications, such as gonadal toxicity. In this case, vaginal obliteration was an early sign of tumor recurrence, although menopause may have contributed to the vaginal obliteration. [*Taiwan J Obstet Gynecol* 2010;49(1):69–71]

Key Words: chemotherapy, cutaneous T-cell lymphoma, Mayer-Rokitansky-Küster-Hauser syndrome, ovarian failure

Introduction

Both transient and permanent ovarian failure is common among women surviving various kinds of hematologic malignancies. Several reports have described the frequency of ovarian failure in women with Hodgkin disease or non-Hodgkin diseases, with rates ranging from 40% to 90% following chemotherapy regimens [1]. Since ovarian failure commonly occurs after chemotherapy, menopause or related sexual activity is often overlooked.

We report a case that emphasizes a clinically rare complication, the complete obliteration of the vagina. The patient died of sepsis and relapse of the underlying malignancy.

Case Report

A 42-year-old married woman had suffered from abdominal pain and spiking fever for days. She had Mayer-Rokitansky-Küster-Hauser syndrome (absence of the uterus, cervix and vagina). A neovagina was established 20 years previously for the purpose of marriage, and she had normal sexual activity thereafter. Sexual activity stopped 1 year prior to this admission, because a cutaneous T-cell lymphoma (stage T4: generalized



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erythroderma [2]) was diagnosed. She was treated with bleomycin, cyclophosphamide, doxorubicin, vincristine, and prednisolone. Before treatment, the gynecologic consultation was unremarkable and the Papanicolaou test was negative for malignant cells. After half a year of treatment, complete clinical remission was achieved, but the woman suffered from severe vasomotor syndromes, including hot flashes, insomnia and night sweating. Hormone profile evaluation showed menopause with an estrogen level of < 20 pg/mL and a follicle-stimulating hormone level of > 30 IU/mL. Neither gynecologic examination nor hormone replacement therapy was performed. She had been in good health and regularly followed up until this admission. At this hospitalization, imaging studies (computed tomography) and laboratory evaluations indicated a suspicious pelvic abscess with leukocytosis ($16,800/\text{mm}^3$) and elevated serum C-reactive protein (18.4 mg/L) levels. Empirical antibiotics, including clindamycin and gentamicin, were prescribed. Surgical intervention and drainage were implemented because of exacerbation of the clinical course, including spiking fever and progressive abdominal pain after a 2-week treatment with antibiotics, and the strong recommendation of infection specialists. Exploratory laparotomy was performed 18 days after admission. This procedure showed severe adhesion in the visceral organs of the abdominal cavity. Random biopsy, including adhesion lysis, and culture were performed. However, the suspicious abscess was not found during operation. The surgical position of the patient was changed from the supine to the lithotomy position to evaluate the deep pelvic cavity. A complete distal vaginal obliteration was noted after identification of the vulva opening. A probe and knife were used to dissect the occluded neovagina bluntly and sharply, and then a Hegar dilator was used to establish the neovaginal channel from the distal end (site of obliteration) to the vaginal stump. A purulent whitish to yellowish discharge flew out from an abscess in the vaginal stump area (proximal end of the reconstructed vagina). A sump drainage system was employed for drainage, and the abdominal wall was closed as usual. Although the patient underwent aggressive medical treatment after surgery, she died 2 months later. Pathology showed widespread lymphoma throughout the perineal area, including the neovagina, deep pelvic cavity and the entire abdominal cavity.

Discussion

Since the introduction of aggressive chemotherapy, alone and in combination with irradiation, long-lasting

remissions and cures have been obtained in patients with hematologic malignancies. Therapy-induced gonadal toxicity has become an issue of clinical concern to these patients [1]. Gonadal injury may not only result in reduced fertility but also affect gonadal steroid synthesis, which may be associated with cardiovascular, sexual and emotional disorders. Instances of successful pregnancy after curing hematologic malignancies have occasionally been reported [3,4]. The use of hormone replacement therapy or oral contraceptives to protect ovarian function is still controversial; however, hormone replacement therapy may be beneficial in young women with therapy-induced menopause [4]. This benefit has yet to be addressed with additional clinical observation. To date, only one reported case has discussed vaginal obliteration after curing a hematologic malignancy [5]. Survival seems to be the priority when physicians manage patients with life-threatening diseases such as hematologic malignancies. However, concern about the frequency and severity of treatment-associated acute and long-term complications such as gonadal toxicity should be emphasized. Management recommendations for these patients should be in accordance with the available evidence. Unfortunately, only rare reports have focused on this aspect of patient management [5–8].

This case of vaginal obliteration may have been caused by chemotherapy-induced gonadal toxicity and a subsequent vaginal defect and occlusion, concomitant lymphoma infiltration of the vaginal wall, or a combination of severe bacterial or fungal infections. We could not determine which factor was the initiator, but menopausal status was important. Of course, severe infection and lymphoma infiltration contributed to these complications.

In summary, physicians should be greatly concerned about the frequency and severity of treatment-associated acute and long-term complications, such as gonadal toxicity. All efforts, including simultaneously using a gonadotropin-releasing hormone agonist [9] should be attempted to minimize the gonad failure-related sequelae [10,11], even if the cancer in the patients has been successfully treated. In this case, vaginal obliteration was an early sign of tumor recurrence, although menopause may have contributed to this problem.

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